

Comprehensive Assessment of Executive Function in Adults with Spina Bifida

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Abstract

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Title: Comprehensive Assessment of Executive Functions in Adults with Spina Bifida

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Objective and method: The study investigates how comprehensive assessment of executive function in adults with Spina Bifida (SB) can provide information of executive dysfunction. In total, 38 patients with SB registered at The National Resource centre for Rare disorders (TRS) participated in the study. As a part of the PhD study "Cognitive rehabilitation in patients with Spina Bifida: Effects of executive functions, psychological and health related factors" (Stubberud, 2010), the participants completed a number of neuropsychological tests, including the Hotel task, as well as self-report questionnaires like the Behaviour Rating Inventory of Executive Function – Adult version (BRIEF-A) and the Hopkins Symptom Checklist -25 (HSCL-25). Pearsons correlational analyses were performed in order to investigate whether the self-report questionnaires and Hotel task could add information on executive dysfunction to the information retrieved from the neuropsychological measures.

Results: Non-significant correlations between BRIEF-A and the neuropsychological tests were found. When controlling for psychological distress measured by HSCL-25, the correlations were nonexistent. This implies that BRIEF-A could provide valuable information on a patient's challenges in daily life, not captured by neuropsychological measures. There were several significant correlations between the Hotel task and the neuropsychological tests, indicating that the Hotel task probably measures some of the same concepts as the neuropsychological tests employed. However, the assessments are not highly correlated. Thus, the implication of the study is that the Hotel task should be included in a comprehensive assessment of executive dysfunction.

Conclusion: The results demonstrated that BRIEF-A and the Hotel task are measures that could be considered for inclusion in a comprehensive assessment of executive function in patients with Spina Bifida.

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1 INTRODUCTION

The study of executive functions is a controversial and complex area within the field of neuropsychology. Several patient groups experience the impact of executive dysfunction on a daily basis. Executive dysfunction often arises as a result of traumatic brain injury or neurological disease. A consequence of the developmental disorder Spina Bifida (SB), is cognitive dysfunction and executive dysfunction in particular. The present study is part of a larger study investigating rehabilitation options for executive dysfunction in patients with SB (Stubberud, Langenbahn, Levine, Stanghelle, Schanke, 2012). There is a large body of research on children with SB, however, there is a dearth of research on adults with SB. In studying measurement of executive function in adults with SB, we are addressing several areas that lack research knowledge.

1.1 Spina Bifida

SB is a developmental disorder caused by a neural tube defect. The defect is a congenital malformation that occurs due to failure of neural tube closure between the third and fourth weeks of gestational age (Frey & Hauser, 2003).

There are various causes of SB, some of these include chromosome abnormalities, single gene disorders, and teratogenic exposures. However, in most cases the cause is not known (Mitchell et al., 2004). At birth, SB seems to be more common among girls than boys with a ratio of 1,2:1 (Foster & Kolaski, 2012). The average incidence of SB worldwide is 1 case per 1000 births, but there are variations in geographical regions as well as periods of time, and by race and ethnicity (Mitchell, et al., 2004). The highest rates are found mainly in Ireland and Wales, with 3-4 cases per 1000 population, and the overall rate is 2-3.5 per 1000 births in the British Isles. In France, Norway, Hungary and Japan, there is a lower prevalence of 0,1 – 0,6 per live births (Foster & Kolaski, 2012). However, estimation of the prevalence of SB has been complicated since the early 1980s in many industrialized countries by the availability of prenatal diagnosis and elective termination of affected fetuses (Chan et al., 1993; Mansfield, Hopfer, & Marteau, 1999).

SB is associated with several abnormalities in the brain, including hydrocephalus and the Arnold-Chiari malformation (Chiari II), meaning a malformation of the cerebellum, with elongation of the cerebellar tonsils. The Arnold-Chiari malformation is also associated with a

smaller sized medulla and pons, as well as internal hydrocephalus (Foster & Kolaski, 2012) . Consequently, SB has a pervasive multisystemic impact on the physical cognitive, psychological, and social functioning of affected individuals (Fletcher et al., 2005). Patients with SB have several areas of characteristic cognitive strengths and weaknesses, likely arising because of impairments in core processes that operate over multiple content domains (Dennis, Landry, Barnes, & Fletcher, 2006).

One study investigated the cognitive status of young adults with SB. A group of the participants had SB with hydrocephalus (SBHC), and another had SB without hydrocephalus (HC). Results showed that the persons with SBHC had a lower cognitive status than the persons without HC. Almost half of the participants with SBHC had cognitive impairments, 70% of these were domain specific, 30% with more general cognitive deficits. The group of participants without HC had a similar cognitive status as a group of healthy participants (Barf et al., 2003). This illustrates the heterogeneity of the display of SB. Some individuals experience cognitive impairments, while others show few or no signs of cognitive impairment.

Another study used magnetic resonance imaging (MRI) to investigate whether the level of spinal lesion in children suffering from SB is associated with variations in anomalous brain development and neurobehavioral outcomes. Participants were divided in a low lesion level group and upper lesion level group regarding the site of the spinal lesion. Compared with the low-level spinal lesion group, the MRIs from children with upper-level spinal lesions showed more qualitative abnormalities in the midbrain and tectum, pons and splenium, and not in the cerebellum. In the latter group, they also found reductions in cerebrum and cerebellum volumes, lower scores on measures of intelligence, adaptive behavior, and a higher frequency of children meeting the criteria of mental retardation. The study concludes that a higher level of spinal lesion is associated with more anomalous brain development, which gives a poorer neurobehavioral outcome in many domains that determine level of independent functioning in everyday life (Fletcher, et al., 2005). Also, Bier and colleagues (1997) administered the Kaufman Brief Intelligence Test (K-BIT) to 65 children and young adults with SB aperta, and found that level of spinal lesion was strongly associated with total K-BIT score . Conclusively, children with SB show anatomical abnormalities in the brain, and this is associated with lower scores on measures of intelligence and a poorer neurobehavioral outcome.

Several studies have found that children with SB especially struggle with tasks involving executive function. Burmeister and colleagues (2005) addressed the incidence of Attention Deficit/Hyperactivity Disorder (ADHD) subtypes in children with SB and shunted hydrocephalus, as well as differences in executive functions in these subtypes. They found that 31% of the group with SB could be identified with ADHD, as rated by parents, and this exceeds the population rate. The group with SB also differed from normal controls on cognitive measures of executive function, but there were no differences between the subtypes.

Results from an early study examining executive processes in children with SB indicated that they had significantly slower processing speed and experienced challenges in problem solving and mental flexibility, compared to normal children as well as children with learning disability and AD/HD (Snow, 1999). Rose and Holmbeck (2007) found that adolescents with SB seem to show a greater impairment on both neuropsychological and self-reported measures of attention and executive functioning compared to healthy controls, even when differences in intellectual functioning were controlled. Further, they argue that the impairment in attention and executive functioning possibly predict their social adjustment difficulties. Another study investigates the relationship between neuropsychological functioning and psychological adjustment in children with SB, measured by the Behavior Rating Inventory of Executive function and The Behavior Assessment System for Children. The study found executive dysfunction to have implications for their psychological adjustment (Kelly et al., 2011). The relationship between SB and neuropsychological functions seems to be well established, however most research is focused on children and adolescents, where adults are not included

In recent years, availability of medical treatment has increased the life expectancy of patients with SB (Fletcher, et al., 2005). Therefore, the question of psychological and neuropsychological functioning with impact on daily living is more focused on adults with SB. The research body is lacking knowledge about the emerging population of people with SB, namely adults. The current study is one of the few focusing executive function on adults with SB. Tarazi, Zabel and Mahone (2008) found that unlike healthy children who have age related improvements in executive control, youth with SB show no such improvements as they grow into adolescence. There is a large body of research implying that youth with SB might face other challenges than the children investigated earlier, such as in academic life, at work, in social relationships and at home, restricting them from participation in society (Barf,

Post, Verhoef, Gooskens, & Prevo, 2010; Barf et al., 2009; Friedman, Holmbeck, DeLucia, Jandasek, & Zebracki, 2009; Heffelfinger et al., 2008; Hetherington, Dennis, Barnes, Drake, & Gentili, 2006; Holmbeck et al., 2010; Stubberud & Riemer, 2011). Therefore, it is likely that the difficulties children and youth face will persist into their adulthood. The challenges adults face might be even more pervasive, as they are not given the same social support as children. As an example, most children have parents that provide care and support. Also, they are often given extra care in school. Adults with SB face higher expectancies and demands on participation in society. In the current study, 53 % of the adult participants reported that they were living alone, 21 % with parents or siblings, only 16 % with own family. Research into how to uncover exactly what areas adults with SB struggle with in daily life, is of great significance. As a consequence, it would be possible to tailor rehabilitation in accordance with a patient's needs.

1.2 Executive function

Executive function is an umbrella term composed of a broad range of behavioral competencies and complex cognitive processes. There is not a consensus on what the term executive functions refers to. Some suggest that executive functions are mainly processed in the prefrontal cortex (Grafman & Litvan, 1999). However, Stuss and Alexander (2000) argues that executive functions are not a unitary concept, but rather distinctive processes related to the frontal lobes, which converge on a general concept of control functions. Further, they delineate that a major problem is an inconsistent and interchangeable use of psychological and anatomical definitions of executive functions and frontal functions. Thus effects of lesions to the frontal lobes has been the common way to understand the frontal lobe functions, yet many still use the term “frontal functions” synonymous to “executive function”, even though there is no objective reference to anatomy (Stuss & Alexander, 2000). Hence, the relationship between “executive” and “frontal lobe” functions are not clear cut, and the anatomical variations complex (Stuss & Alexander, 2000). A main issue when attempting to capture the concept of executive function is the debate on whether or not the frontal lobes can be seen as a unity or comprising a diverse set of functions. Duncan (1995) argued that the executive function is a term representing a unified function of fluid intelligence. On the other hand, others suggest that there are many frontal subfunctions (Shallice & Burgess, 1991; Stuss & Benson, 1984, 1986). Hence, there is controversy in the field of defining what executive functions are like and what areas of the brain are involved.

However, the research body on executive functions has partitioned out a few processes thought to be part of executive functions. Examples of these are labeled “cold” executive functions, such as verbal reasoning, problem-solving, planning, sequencing, ability to sustain attention, resistance to interference, multitasking, cognitive flexibility and ability to deal with novelty (Chan, Shum, Touloupoulou, & Chen, 2008; Stuss, 2011). These functions have been called the “cold” component of executive functions because their corresponding cognitive processes do not involve much emotional arousal, and are not “logically” based. It is suggested that the “cold” components are mediated by the dorsolateral prefrontal cortex (Grafman & Litvan, 1999). The ventromedial prefrontal cortex mediates the “hot” functions, such as obeying the rules of interpersonal social behavior, experience of reward and punishment, and the interpretation of complex emotions (Bechara, Damasio, Damasio, & Lee, 1999; Bechara, Tranel, Damasio, & Damasio, 1996; Grafman & Litvan, 1999). These are components involving emotional arousal. Studies have shown that impairments in the “hot” or “cold” component have significant effects on everyday life, like the ability to work, function independently at home, or maintain social relationships (Goel, Grafman, Tajiik, Gana, & Danto, 1997; Grafman et al., 1996; Green, Kern, Braff, & Mintz, 2000).

Several theories have been developed to explain the cognitive processes belonging to executive functions. Some attempt to connect behavior, mental activity and specific brain regions. Examples of these are Stuss and Benson’s tripartite model (1986), Duncan’s goal-neglect theory (Duncan, 1985) and Damasio’s somatic marker hypothesis (Damasio, 1995). Luria’s theory from 1966 postulates that the human brain consists of three functional units working together. One unit is located in the brain stem and regulates and maintains arousal of the cortex. Another unit is located in the temporal, parietal and occipital lobes, and is responsible for encoding, processing and storage of information. The last unit encompasses functions in the anterior region of the brain and is responsible of programming, regulating and verifying human behavior. Luria considers the prefrontal cortex a superstructure that regulates mental activity and behavior. The latter is a theory driven approach to executive function, and there is a dearth of research connecting the theories based on clinical observations to structural brain anatomy.

The supervisory attentional system model (SAS) of executive functions was proposed by Norman and Shallice (1986), and is an extension of Lurias theory. This model argues that executive functions consist of two systems. One system is responsible for contention

scheduling and the other for supervisory attention. Contention scheduling operates when we perform routine tasks, and helps us prioritize the order of behavior needed to perform these tasks. Sometimes we also need to perform several routine tasks at the same time, for example brushing our teeth while texting an sms, and contention scheduling helps us through this. Supervisory attention is most effective when we are doing novel tasks. Norman and Shallice (1986) give examples on what types of situations are involved in supervisory attention. These are in situations that involve planning or decision-making, situations where we need to correct errors, where responses are novel or not well learned, when we think we are in danger and when we are trying to overcome or resist a strong habitual response or temptation. The SAS model has been extended to include multitasking performance in everyday life (Burgess, 2000; Burgess, Veitch, de lacy Costello, & Shallice, 2000). Eight features of multitasking behavior have been suggested: many discrete tasks for an individual to complete, interleaving period for an effective performance, engagement in only one task at a particular time, interruptions and unexpected outcomes, delayed intentions to return to a task which is already running, tasks that require different characteristics, tasks where one have to decide what is the most appropriate target and performance, and no immediate feedback on the status of performance. Studies on patients with frontal lobe lesions and loss of supervisory control, supports the SAS model (Chan, 2002; Chan, Hoosain, Lee, Fan, & Fong, 2003). Support is also found in studies of healthy subjects demonstrating attention lapses (Chan, 2001; Reason, 1984), and reaction time costs when healthy subjects intentionally switch attention between alternative tasks (Allport, Styles, & Hsieh, 1994; Rogers & Monsell, 1995).

Obviously, there is a considerable amount of controversy linked to the concept of executive function and exactly what this term comprises. Consequently, this leads to challenges in terms of operationalisation of the concept. In the current study, we choose to relate to the SAS model as a framework for understanding the concept of executive function.

Keeping in mind that the term executive function is a controversial one, it is still possible to infer what an executive dysfunction could be. Myiake et al (2000) suggests that among unitary executive processes we have mental set shifting, information updating and monitoring and inhibition of reflexive responses. An example of executive dysfunction could be when these functions are not working properly, as may happen due to brain damage or neurological disease. It has been commonly assumed that executive dysfunction implies lesion of the frontal lobes and this has often been inferred from results on neuropsychological tests.

Planning and execution has been mentioned as a part of the “cold” executive functions. Patients with lesions of the dorsolateral prefrontal cortex (DLPFC) often have severe difficulties with planning and execution (Dubois, Andrade, & Levy, 2008). The patients show a decreased ability for conceptualization and attention, an impaired ability to activate cognitive strategies (word-finding difficulties, impaired memory retrieval), difficulties in set-shifting and distractibility when performing complex cognitive tasks. Consequently, they have problems finding a rule, shift mental sets, solve multiple-step problems and retrieve information (Dubois et al., 2008). Problems with planning and execution could give severe challenges on a daily basis. Another study found that patients with focal frontal lesions performed as well as control participants on measurements of dual capacity and ability to inhibit irrelevant information, despite showing reduced short-term storage. This could imply that not all executive processes are exclusively sustained by the frontal cortex (Andrés & Van der Linden, 2002). Goal directed behavior has been termed as part of executive function. This implies an ability to initiate behavior, control impulsivity, capacity to perceive congruence between the goal and the consequences of the projected behavior, and the sensitivity to reward. Deficits in behavioral activation may be caused by lesions of the orbital and ventromedial parts of prefrontal cortex (Dubois et al., 2008). Patient E.V.R. as described by Eslinger and Damasio (1985) had severe frontal lobe lesions. He seemed to have no cognitive deficit as measured by neuropsychological tests, yet he presented with severe impairment in goal-directed behavior. Patients such as E.V.R. may experience difficulties evaluating the consequences of their behavior (Dubois et al., 2008). The orbital and ventromedial parts of the prefrontal cortex are areas normally involved in this type of behavior, affecting sensitivity to the affective value of behaviors and anticipation of reward (Rolls, 1990). As a consequence, this could present in the ability to develop and maintain social relationships as well as the ability to meet expectations at work or school.

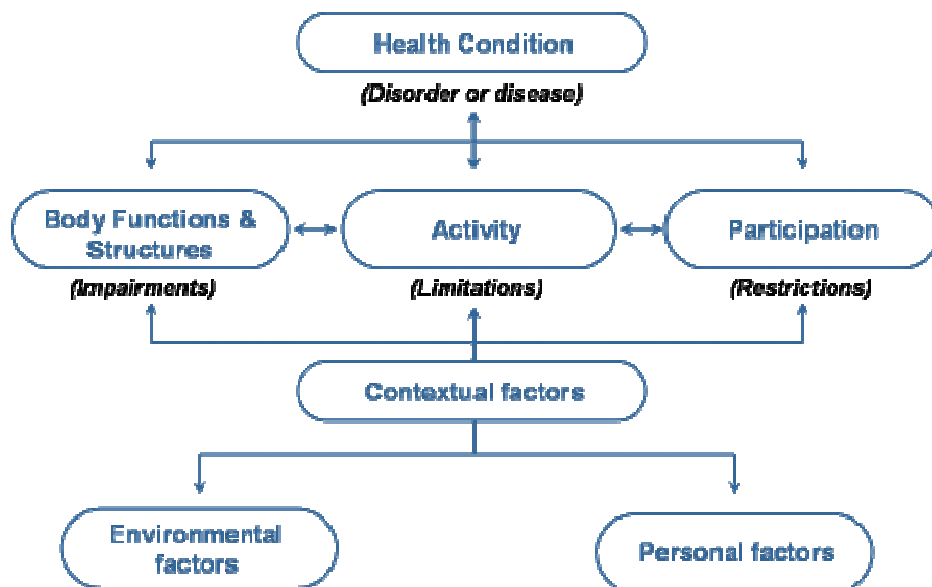
It is clear that the term executive function and the localization of these functions anatomically are not completely agreed on. Consequently, the understanding of executive dysfunction is equally confusing. Yet, it is well established that executive dysfunction can lead to disruption in everyday living, even when other cognitive functions are relatively intact (Damasio, 1994). Executive dysfunction may cause a number of behavioral and psychosocial challenges, like dealing with novel situations, problems forming a reasonable plan that takes the relevant details into account, inhibiting responses to situations when these are

inappropriate and increased distractibility (Levine et al., 2007). These impairments are handicapping in the social and occupational domains of life.

1.3 International Classification of Functioning, Disability and Health

The International Classification of Functioning, Disability and Health (ICF) developed by World Health Organisation (1980, 2001) gives descriptions of functionality levels relating to the consequences of disease. ICF is a revised version of the Classification of Impairment, Disability and Handicap (ICIDH), and includes social aspects of disability, not viewing it only as a medical or biological dysfunction. The ICF model is illustrated in Figure 1. There are different levels describing how a disease might affect a person. The impairment level describes a loss or abnormality of body structure of a physiological or psychological function (World Health Organization, 2001). This perspective describes a view of the illness within a person. The activity level refers to the degree to which a person with an illness is able to carry out different activities of daily living due to a disability. It describes the degree to which a person experiences limitations in activities due to an illness. The last level is participation and relates to how well a person can participate in various situations of life. It incorporates impairments, activities and health condition with the context a person lives in. Degree of participation is the result of an interaction between all the beforementioned factors. A participation reduction means a disadvantage in social, familial and educational roles in relation to a disease. In conclusion, impairment level is concerned with the illness within a person, while activity and participation relates to the external consequences of the impairment. These three levels are all affected by external factors such as availability of rehabilitation options, economy, social support. Also, personal factors are taken into consideration, such as age, gender, general fitness and cultural background. All of these aspects of a person and a disease are incorporated into a broad perspective of a persons health status.

Figure 1. The ICF model



The ICF model is not a measure of severity of disease by itself, but a framework for understanding the consequences of disease. Recently, the use of this framework within neuropsychological rehabilitation has gained attention. It has been recommended to use the framework in developing a broad understanding of the challenges patients struggle with, especially in the context of daily life (Lewis, Babbage, & Leathem, 2011; Wilson, 2008). This places new demands on how to assess executive dysfunction, and will be discussed further in 1.4.

1.4 Assessment of executive functions

To assess level of executive function and possible impairment it is common to use a neuropsychological battery of tests including, for example, the Wechsler Adult Intelligence Scale, and subtests from the Delis-Kaplan Executive Function System (D-KEFS; Delis, Kaplan & Kramer, 2001) such as the Color-Word Interference test and Trail Making Test.

The SAS model proposes different systems involved in executive functions. There is contention scheduling operating when we perform routine tasks and supervisory attention is in action when we perform novel tasks. Supervisory attention could be operating when we assess executive functioning in a laboratory with neuropsychological tests, because these tests

propose novel challenges to a testee. Chan et al., (2008) suggests that the D-KEFS Tower Test (Delis et al., 2001) and the Hotel task (Manly, Hawkins, Evans, Woldt, & Robertson, 2002), among others, are relevant for testing executive function in line with the SAS model, tapping planning and strategy allocation. However, as the Hotel task and neuropsychological tests are most likely to present novel situations to the patient, they possibly lack the ability to tap the function of contention scheduling. However, also during our daily routines either at home or work, new situations show up that we need to handle and incorporate into our routine problem solving. This could mean that supervisory attention is also relevant on a day to day basis, working together with contention scheduling. Either way, contention scheduling is relevant on a day to day basis, and also needs to be assessed to understand a testee's executive function as a whole.

Chan et al. (2008) suggests that there are a few obvious difficulties in the neuropsychological study of executive deficits and its rehabilitation. One of them is what he calls a fractionation of the executive symptoms. How a patient performs on one test of executive function could have little or no predictive value of performance in a complex real world situation. Therefore, we see an increased emphasis on using more complex and life-like measures as an add-on to neuropsychological tests. These measures would investigate the functioning of several executive domains at the same time and give additional information on a patient's level of impairment, activity and participation according to ICF. In line with this, some argue that neuropsychological tests do not consistently pick up on the difficulties a patient might experience in his or her daily life (Torralva, Roca, Gleichgerricht, Bekinschtein, & Manes, 2009), namely their disability. Eslinger and Damasio (1985) epitomize the challenge of only assessing the impairment level of ICF with a frontal patient called E.V.R. The patient had average to superior scores on impairment tests of executive functioning, but still had severe difficulties in daily tasks involving planning, decision-making and judgment (Eslinger & Damasio, 1985). This gives an indication that assessment of executive functions is different from measuring other cognitive abilities, meaning that we are dealing with heterogeneous concepts and that executive dysfunction is difficult to tap. Additional studies have also shown that many patients with executive problems in daily life perform well on tests assumed to be sensitive to executive functions, while still other studies have found results to the contrary (Cripe, 1998; Eslinger & Damasio, 1985; Shallice & Burgess, 1991). One explanation for this could be that neuropsychological tests are administered in a controlled environment without disturbances and distractions, unlike a natural daily living environment. In a natural

environment the patient is not protected against distractions competing for attention like in a test situation. This is one of the most problematic areas of assessment of executive functioning (Lezak, 1982). Consequently, there is often a weak to moderate relationship between performance on neuropsychological assessments and everyday behavior (Manchester, Priestly, & Jackson, 2004). Some argue that in formal neuropsychological testing, patients are not faced with noticing whether or not a task needs to be done, and they are not required to choose a task from a number of competing tasks. The affective arousal such situations may cause is controlled for, and consequently a variable of huge importance to decision making and behavioral control in everyday life is omitted from the assessment process (Manchester, et al., 2004). There are few real world situations in which neuropsychological measures are analogue (Burgess et al., 2006). Therefore, a patient might not show severe difficulties and signs of impairment on a neuropsychological test, and still experience challenges when the same executive functions are challenged at home or in the working place. In some populations, for example patients with traumatic brain injury, children with ADHD and patients with frontotemporal dementia, it seems the neuropsychological test battery might leave a false or partial impression of neuropsychological functioning in daily life (Gioia, & Isquith, 2004; Mcauley, Chen, Goos, Schachar, & Crosbie, 2010; Toplak, Bucciarelli, Jain, & Tannock, 2009; Torralva, et al., 2009; Vriezen & Pigott, 2002). Could the same be true for adults with SB?

The traditional neuropsychological tests have been developed for diagnostic purposes, related to the impairment level of ICF. This may be another reason why such tests cannot consistently predict everyday behavior, related to the activity and participation level of ICF. Recent development however, has seen a switch from diagnostic purposes only to an emphasis on ecological validity, that allows the measurements to predict social and functional abilities in a real life setting (Wood & Liossi, 2006). This is important in relation to the new development in WHO's standard of analysing consequences due to illness. Lewis and colleagues (2011) argue that for neuropsychological assessments to be useful for cognitive rehabilitation, the assessments should be able to predict functioning in everyday life. This means that the measures needs ecological validity. Ecological validity has been termed as an ability to generalize results of controlled experiments to naturally occurring events in the real world (Brunswick, 1955). Verisimilitude and veridicality are two concepts on which ecological validity of tests rely (Chaytor & Schmitter-Edgecombe, 2003), and it relates to neuropsychological testing. Verisimilitude reflects the degree to which a test resembles the

cognitive demands of a real life task. Veridicality relates to the degree to which performance on a neuropsychological test is empirically related to everyday functioning (Wood & Liossi, 2006). Looking to the ICF framework this may give direction to where the field of assessment in psychology could go, not only focusing on locating a lesion through neuropsychological tests, but also strive to understand what symptoms a patient lives with on a daily basis. A person's cognitive capacity in a standardised environment, as seen when using neuropsychological tests, can only tell us what that person can do in a structured environment, and not predict the person's typical functioning in a day-to-day environment (Chaytor & Schmitter-Edgecombe, 2003). This challenges the scope of neuropsychological testing.

1.5 Behavior Rating Inventory of Executive Functions Adult Version

The Behavior Rating Inventory of Executive Functions – Adult (BRIEF- A) is a self-report questionnaire constructed to measure executive functioning in daily life situations with ecological validity in mind (Gioia & Isquith 2004). The original version was developed for children, and the adult version was derived from this. The questionnaire has gained clinical popularity the last decade (Løvstad, Endestad, & Funderud, 2010). Nevertheless, there is a dearth of research on the relation between BRIEF-A, and neuropsychological measures on executive function, especially in the adult population.

To investigate the concurrent validity of BRIEF, Mahone and colleagues (2002), compared results from the BRIEF with additional behavior rating scales and performance based measures of executive function in children with AD/HD and/or Tourette syndrome. They found that the BRIEF index scores showed no significant correlation with performance based measures of executive functions. However, there was a strong relationship between BRIEF, interviews with parents and other rating scales measuring behaviors seen in AD/HD. One study compared test results on BRIEF-A and neuropsychological tests from patients with focal lesions to the orbitofrontal (OFC) and lateral prefrontal cortex (LPFC) (Løvstad, et al., 2010). It was found that BRIEF-A was more sensitive to detect problems in everyday living in patients with OFC lesions, than in patients with LPFC lesions. The LPFC group displayed more problems on the neuropsychological tests than on BRIEF-A. On the other hand, the neuropsychological measures did not detect the challenges of patients with OFC lesions.

Some studies have found a high correspondence between self-rate measures and neuropsychological tests (Basso et al. 2008; Marrie, Chelune, Miller, & Cohen, 2005).

However, in other studies, the correspondence between self-report measures and neuropsychological tests have not been found (Benedict et al., 2003; Middleton, Denney, Lynch & Parameter, 2006). Psychological factors such as depression or coping mechanisms are suggested to relate to overestimation of cognitive dysfunction on subjective measures such as the BRIEF-A (Julian, Merluzzi & Mohr, 2007; Maor, Olmer & Mozes. 2001; Gold, Schulz, Mönch, Schulz & Heesen, 2003). On the other hand, overestimation of cognitive performance is suggested to relate to fewer symptoms of depression, conscientiousness and greater degree of cognitive impairment (Carone, Benedict, Munschauer, Fishman, & Weinstock-Guttman, 2005). The abovementioned factors such as coping mechanisms and depression could contribute to the discrepancy in results from self-report questionnaires and neuropsychological tests (Gold et al., 2003).

The BRIEF-A includes an informant questionnaire in addition to the self-rating form. This could help circumvent the problems many patients with executive dysfunction have with accurately reporting their difficulties (Manchester et al., 2004). Despite this, there are also a few challenges to the accuracy of collateral information. These may be the informants familiarity with the patient's everyday functioning (Norris & Tate, 2000), family stress and dynamics (Fleming, Strong & Ashton, 1996), time since injury (Chevignard et al., 2000), effects of stereotypical views of patients performance held by some clinicians (Cavallo, Kay & Ezrachi, 1992), and rigid perceptions of the patient from relatives and staff, even when the condition of the patient later improves (Sohlberg, Mateer, Penkman, Glang, Todis, 1998).

In conclusion, this implicates that the behavioral difficulties due to executive dysfunction in some patients may not be easily discovered through neuropsychological testing alone. It also points to the difficulties in assessing executive dysfunction, demonstrating that the SB group of patients is highly heterogeneous. A suggestion is that for some groups of patients the BRIEF-A could be more sensitive than neuropsychological tests, and shed additional light on their difficulties. Yet, we cannot look past the question on whether or not BRIEF-A actually assesses executive function, or whether it is more sensitive to psychosocial stress in general, as experienced by some patients with executive dysfunction.

1.6 Hotel task

The Hotel task is designed to measure planning, strategy allocation, ability to switch between tasks (Chan, et al., 2008; Manly, et al., 2002) and measure performance on “daily life” activities in a “real life” environment (Torralva, et al., 2009). However, few studies have so far examined the validity of the Hotel task.

One study investigated the usefulness of incorporating the Hotel task in assessment batteries, to tap specific cognitive deficits in patients with bipolar disorder (BD) (Torralva et al., 2012). Torralva et al., (2012) argues that a subgroup of BD patients performs within the normal range on neuropsychological tests, even though a growing body of evidence suggests that they are likely to have cognitive deficits. Torralva et al. (2012) advocates that this could be due to a lack of ecological validity in the standard neuropsychological battery. To test their hypothesis, a control group and a BD group completed a standard neuropsychological test battery including the Hotel task among other tests. They found that the BD group had comparable results to the control group on standard neuropsychological tests, but showed deficits in executive functioning when completing the Hotel task. Based on this, Torralva et. al (2012) conclude that the inclusion of measures such as the Hotel task could result in a more realistic picture of the patients functioning as a whole, thereby easing the design of therapeutic and rehabilitation strategies. However, it is also worth questioning whether the affective profile of patients with BD could have been picked up by the Hotel task, thereby masking their true cognitive abilities.

Torralva and colleagues (2009) have incorporated the Hotel task in a test battery of Executive and Social Cognition (ESCB). In their study they investigated whether ESCB was sensitive for early detection of executive and social cognitive impairments in a group of patients with frontotemporal dementia. They divided their participants into two groups, according to their scores on Addenbrooke’s Cognitive Examination (ACE). Significant differences were found between the low ACE group, and the healthy control group on neuropsychological tests and the Executive and Social Cognition Battery. However, the participants in the high ACE group did not differ from the control group on neuropsychological tests. Nevertheless, significant differences were found between the high ACE group and the controls on ESCB scores, including scores on the Hotel task. These results suggest that the Hotel task, as incorporated in the ESCB could be more sensitive in detecting executive and social cognitive impairments than neuropsychological tests.

In conclusion, there is some evidence supporting the validity of Hotel task as a measure of cognitive abilities. Even so, this could be investigated further, especially with ecological validity and executive functions in mind.

1.7 Purpose of study

Knowing that adult individuals with SB are likely to display executive deficits and possibly problems in psychosocial adjustment and daily living, it is important to understand how to tap into and shed light on their difficulties. In this study, the SAS model will be used as a framework for understanding executive function. Based on this, the investigation will make use of neuropsychological tests developed for tapping diverse aspects of executive function in line with the theoretical model of SAS. When measuring executive deficits we would argue that assessment should be tailored towards uncovering consequences due to executive dysfunction in line with the ICF framework, in order to attain a broader perspective of a patient's difficulties and rehabilitation needs. Hence, in addition to neuropsychological tests, this study includes the Hotel task and a self-report questionnaire focused on day-to-day executive behavior (BRIEF-A). We hypothesise that the Hotel task and BRIEF-A add information to neuropsychological performance based measures, the latter assessing deficits on the impairment level according to ICF. In line with ICF we hope firstly to broaden the understanding of the problems those with SB are dealing with, and secondly to give recommendations for treatment tailored to their specific needs with consequences for participation in daily life, work and social relationships. According to Wilson (2008), failure to identify those patients who can and cannot be helped creates a lack of credibility in rehabilitation. The objectives of this thesis are the following:

Objective 1: BRIEF-A was developed to measure of executive function in daily life. Previous research on children and adolescents with traumatic brain injury has found weak positive correlations between performance on standardised tests and executive functions in daily life assessed by BRIEF. (Anderson, Anderson, Northam, Jacobs, & Mikiewicz, 2002; Vriezen & Pigott, 2002). Therefore, an objective of this study is to investigate if the same pattern can be found with adults with SB. If, as expected, low positive correspondence correlations are found, it could mean that BRIEF-A and the performance based measures tap different levels of executive function.

Objective 2: It has been suggested that the relationship between subjective and objective measures of executive function could be confounded by psychological factors such as symptoms of depression (Julian et al., 2007; Maor et al., 2001). One study found that children and adolescents with SB show a higher degree of internalising symptoms, adaptive functioning and depressive symptoms (Kelly et al., 2011). Hence, the second objective of this study is to investigate whether scores of psychological distress as measured by HSCL-25 confounds the relationship between BRIEF-A ratings and scores on performance based measures. This would provide an alternative or additional explanation to the expected relationship between BRIEF-A and neuropsychological measures.

Objective 3: The third objective of this study is to compare performance based test results with those of the Hotel task. Based on previous research on other patient groups (Torralva, et al., 2012), it is hypothesized to find only weak positive correlations between the Hotel task and the various neuropsychological tests. Also, in line with the SAS model, the Hotel task and the neuropsychological tests could relate to different parts of executive function, which leads to an expectation of low to moderate correspondence between them. This could mean that different methods of measurement tap on different levels of executive function, namely impairment, activity and participation as described in ICF. If the results are as predicted, it strengthens the argument that a comprehensive approach in assessing executive dysfunction is needed in the rehabilitation setting.

Objective 4: The fourth objective of this study is to investigate the relationship between BRIEF-A and Hotel task. The former being a questionnaire tapping day to day executive function, the latter being a laboratory test trying to incorporate several aspects of a realistic situation in day to day life. According to Chan et al. (2008) the Hotel task is likely to tap activity level, whereas the BRIEF-A taps both participation and activity level of ICF, inclining that to gain insight to the different functionality levels, both types of tests should be used. Consequently it would be expected that the Hotel task and BRIEF-A show few significant correlations.

Combined, this could have implications on how to assess level of impairment and disability in adults with SB. In line with arguments proposed by Chan et al. (2008), best practise of assessment should be based on a comprehensive view of the patient's overall function, thereby giving direction to how rehabilitation should be planned and realised.

In summary; we raise these research hypotheses:

1. BRIEF-A and neuropsychological measures of executive function does not tap the same level of executive function in line with the ICF model.
2. An elevated level of psychological distress as measured by HSCL-25 will affect how executive dysfunction is reflected through BRIEF-A and neuropsychological measures.
3. The Hotel task and neuropsychological measures of executive function does not tap the same level of executive function in line with the ICF model.
4. Hotel task and BRIEF-A does not tap the same level of executive function in line with the ICF model.

2 METHODS

The study will use data collected for the study “Cognitive rehabilitation in patients with Spina bifida: Effects of executive functions, psychological and health related factors” (Stubberud, 2010).

2.1 Participants

Participants in the study are a group of patients diagnosed with SB (N=38). Mean age of the participants was 31,8 years. There were 16 males and 22 females. Twenty-six percent had completed primary school, 61 % upper secondary and 5 % had completed higher education. Seven of the participants were married. Twenty of them were living alone, the rest with own family, parents, siblings or other.

Figure 2. Sample characteristics

	Total (N=38)
Age, mean \pm SD	31.79 (8.31)
Gender (M=men, F= female)	16 M, 22 F
Education, years \pm SD	12.3 (1,78)
- Primary n (%)	10 (26)
- Upper secondary n (%)	23 (61)
- Higher education n (%)	5 (13)
Marital Status (with partner) n (%)	7 (18)
Paid work full time n (%)	3 (8)
Living situation n (%)	
- Living alone	20 (53)
- With parents/siblings	8 (21)
- Own family	6 (16)
- Other	4 (11)
Hydrocephalus n (%)	33 (87)
- Shunt	31 (82)
- 3 rd ventriculostomy	4 (11)
- > 3 shunt revisions	15 (39)

2.2 Procedure

In the PhD study “Cognitive rehabilitation in patients with SB: Effects on executive functions, psychological and health related factors”, patients between 20 and 45 years of age registered at The National Resource Centre for rare disorders, where asked to participate (N=201). In the information letter, BRIEF-A was attached to fill out and returned in order to screen for executive problems. Those who responded and obtained the score $T \geq 60$ on the Metacognition Index on BRIEF-A were included in the study (N=53). The metacognition score is thought to tap an individual’s ability to initiate activity, generate problem-solving ideas, sustain working memory, plan and organize one’s materials (Gioia, Isquith, Guy, & Kenworthy, 2000). For those included, further screening measures were included. In this study, baseline measurements on BRIEF-A and the neuropsychological tests will be used for further analysis. For a description of those included and excluded from the study, see flow chart in figure 3. The following were used as inclusion and exclusion criteria:

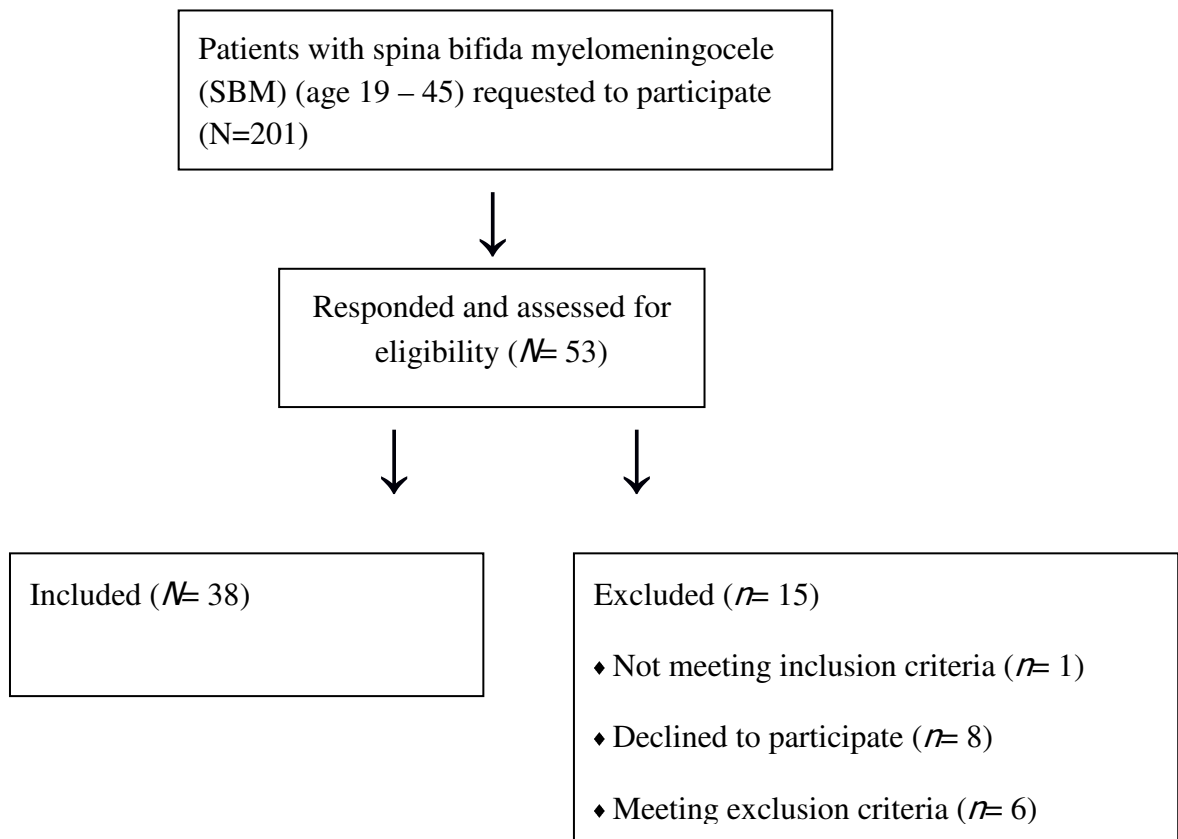
Inclusion:

- SB with myelomeningocele (MMC)
- Between 20 and 45 years of age
- Problems in the domain of executive functioning assessed by BRIEF-A

Exclusion:

- Major psychiatric disorder on Axis I psychiatric disorders, screening was done based on the patients journal
- Reported alcohol or substance abuse within the past year
- Aphasia or other specified language problems causing potential validity problems
- $IQ < 70$

Figure 3. Procedure



2.3 Ethics

The project is conducted in accordance with guidelines by the Helsinki declaration and Vancouver rules. The PhD study is approved by the Regional Ethic Committee for Medical Research Ethics, South-Eastern Norway and the Norwegian Data Inspectorate. The data selected for use in this study are anonymous by the hand of the student.

2.4 Materials

Self report measures

1. Behavior Rating Inventory of Executive Functions – Adult Version (BRIEF-A; Gioia et al., 2000). A Norwegian translation of the self measurement form was used. The BRIEF-A is a questionnaire with 75 questions. The questions can be answered on a scale rating whether a stated behavior is experienced as problematic “never”, “sometimes” or “often”, the last six months. A T-score is calculated based on the responses, and for a score to be considered elevated on the BRIEF-A, it has to be higher than $T \geq 65$ (Gioia et al., 2000).

There are two rating forms, one self-measurement form and one informant form. In this study only the self-measurement form was used. The BRIEF-A has two indexes composed of more clinical subscales. The Behavioral Regulation Index (BRI) consists of subscales Inhibit, Shift and Emotional Control. The Metacognition Index (MI) consists of Initiate, Working Memory, Plan/Organize, Organization of Materials and Monitor subscales. The Global Executive Composite (GEC) score incorporates all clinical scales. All of the indexes were included in the analyses for this study. Higher scores on the indexes indicate greater executive dysfunction. There are two validity scales measuring Negativity and Inconsistency of Responses.

2. Hopkins Symptom Check List 25 (HSCL-25; Derogatis, Lipman, Rickles, Uhlenhuth, Covi, 1974). Norwegian translation. HSCL-25 is a questionnaire consisting of 25 questions. The respondents are asked to consider how often they have been bothered by symptoms such as headache, dizziness or sudden fear during the last seven days, on a scale from 0-4. The questionnaire has three indexes, one for anxiety, one for depression and a global scale incorporating the two former scales. Responses on question number 1-10 in the questionnaire are used to calculate the anxiety index. Responses on question number 11 – 25 are used to calculate the depression index. All responses are used for a global index of psychological distress. There is no established cut-off score for HSCL-25. However, for the scale used in this study (0-4), a mean index of $\geq 1,0$, is considered a sign that the respondent has as much symptoms of anxiety and/or depression as traditional psychiatric patients diagnosed with anxiety or depression (Ravndal & Amundsen, 2010). Hence, higher scores indicate more symptoms of depression or anxiety. To calculate the mean index for anxiety, scores on questions 1 - 10 are averaged. The index for depression is the average of responses on questions 11 – 25. Both the anxiety and the depression index were used in this study.

Objective measures of cognitive functioning

3. D-KEFS Tower test, (Delis et al., 2001). This test is used to measure planning ability, working memory, response inhibition and visuospatial memory (Carlin, Bonerba, & Phipps, 2000; Phillips, Wynn, & Gilhooly, 1999; Welch, Revilla, Strongin, & Kepler, 2000). The test consists of asking participants to complete tower building tasks by recreating pictured constructions using an increasing number of disks placed on three spindles. Tests are timed, are of increasing complexity and require planning and

flexibility to complete the tasks as quickly as possible. The materials are 5 disks that vary in size, and a board with three vertical pegs. In order to perform the task as fast as possible and without breaking any rules, the participant needs to plan his/her moves ahead. The parameter included in this study was total achievement score.

4. D-KEFS Trail Making Test (TMT). TMT is a pen and paper task, and it involves connecting numbers and letters sequentially without removing the pencil's tip from the paper. The test has traditionally been used to measure visual scanning and visuomotor tracking, divided attention and cognitive flexibility, for example in shifting from letter to number in condition 4 (Lezak, Howieson, & Loring, 2004). Motor speed also contributes to success on this test (Schear & Sato, 1989). The D-KEFS version of TMT consists of five conditions, but only results from condition four was included in this study. Condition four corresponds directly to part B of the traditional TMT from the Halsted-Reitan Neuropsychological Test Battery. Performance on the test is timed and the time variable is used in this study.
5. D-KEFS Color-Word Interference Test (CWI). CWI measures response inhibition and cognitive flexibility (Lezak, et al., 2004) . The test includes four different conditions and in this study conditions three and four was used. The parameters for conditions three and four were time spent to complete the test and errors made and corrected by the participant (corrected, uncorrected and total).
6. Conners Continuous Performance Test II (CPT-II; Conners, 2000). Measures the ability to sustain and focus attention. This computer based test takes 14 minutes to complete. The task demands concentration because it is somewhat monotonous (Lezak, et al., 2004). The respondent is asked to hit a button every time a target letter appears on a screen. There are several measures available from the CPT II and the following are examined in this study: omissions, commission, detectability and perseverations. Omissions result from a failure to respond to target letters. Commission errors are made when responses are given to non-target letters. Detectability is a measure of the difference between the amount of targets correctly hit and amount of non-targets hit. This gives an idea of the participant's discriminative power. Perseverations are any reaction less than 100 ms. Such a response indicates either a slow response to a preceding stimuli, a random response, anticipatory response or random response.

7. The Hotel Task (Manly et al., 2002). Norwegian translation. In the Hotel task participants are asked to do 6 sub-tasks within 15 minutes. The tasks are compiling individual bills, sorting the charity collection of coins, looking up telephone numbers, sorting conference labels, and proof reading the hotel leaflet, as well as opening and closing garage doors at set times. The participant has a watch at hand to keep track of time. He or she is asked to try out every one of the tasks within 15 minutes, just to get a sense of them. The participant is not supposed to finish all the tasks, just try out each of them and spend as much time as possible on each during the time allocated. All tasks can be completed in what order the participant choose, except for the garage door, which needs to be opened and closed at set times. Performance on the Hotel task is scored within several categories described in the following. The participant is given one point for each task attempted. Participants are asked to spend as much time as possible on each task and the optimal time spent is consequently three minutes per task. Deviations from this is calculated for each of the tasks and summed up. One point is given for pressing each of the garage door buttons. Since the participants are also asked to press the garage door buttons at particular times, deviations from this is calculated in seconds.
8. Wechsler Abbreviated Scale of Intelligence (WASI; Wechsler, 1999). WASI was developed as a short form of measuring intellectual functioning and is composed of four subtests, two verbal and two performance (Axelrod, 2002). In this study, the full scale IQ score (FSIQ) is used to provide an impression on the participants general ability level. FSIQ is generated using results from all four subtests.

3 RESULTS

3.1 Statistics

All the scores on the neuropsychological tests, BRIEF-A and Hotel task were inspected for normality and outliers. The data was normally distributed with no disturbing outliers.

Therefore, no cases were excluded from the further analyses. Analyses were conducted using PASW Statistics 18.0 software (SPSS Inc.). Correlational analyses were estimated using Pearsons product-moment correlation. Partial correlations and stepwise regression analyses were estimated for selected analyses.

3.2 Descriptive statistics

Descriptive statistics of the neuropsychological data are presented in Table 1.

Table 1. *Descriptive statistics of neuropsychological data (N=38)*

Variable	\bar{X}	SD	Min – Max
WASI FSIQ	91,52	14,10	70 - 121
BRIEF-A			
- Global Executive Composite	64,10	10,51	46 - 87
- Metacognition Index	66,44	9,51	48 - 88
- Behavioral Regulation Index	58,71	12,31	38 - 81
HSCL25			
- Depression	18,50	13,21	2 - 49
- Anxiety	7,36	6,30	0 - 35
- Global Symptom Index	25,94	17,57	3 - 80
Hotel task			
- Attempted tasks	3,63	1,24	2 - 5
- Total time deviation (sec)	680,65	295,08	226 - 1176
- Total time deviation garage doors (sec)	386,84	450,44	0 - 1080
CPT			
- Omission	5,71	12,37	0 - 68
- Commission	19,39	7,30	3 - 34
- Perseveration	0,78	1,33	0 - 5
- Detectability	0,37	0,34	-0,28 –1,3
Tower test			
- Total achievement score	12,68	6,21	2 - 23
- TMT (condition 4)			
- Raw time (sec)	122,92	52,00	61 - 240

- Set loss errors	0,36	0,59	0 - 2
<hr/>			
CWI (condition 3)			
- Time (sec)	71,47	19,40	46 - 115
- Corrected errors	1,21	1,54	0 - 7
- Uncorrected errors	0,28	1,08	0 - 6
- Total errors	1,5	1,73	0 - 7
<hr/>			
CWI (condition 4)			
- Time (sec)	88,28	28,43	48 - 162
- Corrected errors	1,26	1,32	0 - 6
- Uncorrected errors	1,39	3,24	0 - 19
- Total errors	2,68	3,79	0 - 21

NOTE: All scores reported are raw scores, except the scores on BRIEF-A and HSCL-25, which are reported in standardized T-scores, and WASI which is reported in Intelligence Quotient. FSIQ WASI = Full Scale Intelligence Quotient Wechsler Abbreviated Scale of Intelligence, BRIEF-A = Behavior Regulation Index of Executive Function – Adult version, HSCL25 = Hopkins Symptoms Checklist 25, CPT = Conners Continuous Performance test II, TMT = Trail Making Test, CWI = Color-Word Interference test
N=38

Participants scored an average of 91, 5 on FSIQ, which is somewhat lower than the average score of the general population, which is 100. The highest FSIQ score in the group was 121, and the lowest was 70. The cut off for participating in the study was 70, so any individuals scoring lower than this were not included.

For a score to be considered elevated on the BRIEF-A, it has to be higher than $T \geq 65$ (Gioia et al., 2000). Our group of participants show on average elevated scores on the GEC and MI, but not the BRI. The statistic material has no significant outliers precluding the mean score provided here.

As mentioned, there is no established cut-off score for HSCL25. However, as suggested by Ravndal and Amundsen (2010), a mean index of $\geq 1,0$, is considered a sign that the respondent has as much symptoms of anxiety and/or depression as traditional psychiatric patients diagnosed with anxiety or depression. Our group of participants most likely experiences a notable amount of symptoms of depression, but not anxiety. This is calculated using the following procedure. For the anxiety index, the mean scores for all participants were used and divided with number of questions related to anxiety in the HSCL25. This would be 0,73 (7,3/10). For the depression index, it would be 1,2 (18,5/15).

3.3 Relationship between BRIEF-A and neuropsychological tests

In order to examine whether BRIEF-A and the performance based measures tap different or similar levels of executive function (objective 1), correlations between BRIEF-A and neuropsychological tests were estimated using Pearsons product-moment correlation. The most relevant results are presented in Table 2.

Table 2. *Correlations between BRIEF-A and neuropsychological tests*

Variable	BRIEF-A		
	Global Executive Composite	Metacognition Index	Behavioral Regulation Index
Tower test			
- Total achievement score	-,261	-,173	-,312
TMT (condition 4)			
- Raw time (sec)	,355*	,361*	,303
- Set loss errors	,111	,008	,201
CWI (condition 3)			
- Time (sec)	,143	,219	,055
- Uncorrected errors	,240	,102	,345*
- Corrected errors	-,021	,003	-,032
- Total errors	,132	,066	,188
CWI (condition 4)			
- Time (sec)	,467**	,439**	,417**
- Uncorrected errors	-,087	-,064	-,086
- Corrected errors	,240	,225	,231
- Total errors	,011	,026	,008
CPT			
- Omission	,052	,065	,034
- Commission	,145	,120	,136
- Detectability	-,109	-,094	-,095
- Perseverance	,105	,141	,068

NOTE: Scores used for the analyses were raw scores, except scores from BRIEF-A which were standardized T-scores. BRIEF-A = Behavior Regulation Index of Executive Function – Adult version, CPT = Conners Continuous Performance test II, TMT = Trail Making Test, CWI = Color-Word Interference test. * $p < 0.05$ ** $p < 0.01$, N=38

It can be seen from the data in Table 2 that the correlations between BRIEF-A and the various neuropsychological tests are of quite different magnitude, ranging from zero to a correlation as strong as 0,467 ($p<0,01$). The correlations are in general in the expected direction, i.e. high scores on BRIEF-A is related to poor performance on the various tests. There are, however, few significant relationships discovered; MI has a significant positive correlation with time spent on TMT (condition 4) and time spent on the Color-Word Interference test (CWI) (condition 4). The BRI correlates positively time spent on CWI (condition 4). Though not significant, there is also a positive correlation between BRI and time spent on TMT (condition 4), as well as Tower total achievement score.

MI and BRI are composite indexes of several different scores, and so it would be interesting to look further into exactly what subscales that correlates with the neuropsychological tests. The correlations are presented in Table 3 and Table 4. CPT is taken out of the further analysis because no significant relationship was found between the test and BRIEF-A.

Table 3. *Correlations between Behavior Regulation Index subscales and neuropsychological tests*

Variable	Behavior Regulation Index Subscales			
	Inhibit	Shift	Emotional Control	Self Monitor
Tower				
-Total achievement score	-,253	-,280	-,222	-,340*
TMT (condition 4)				
-Time (sec)	,363*	,358*	,141	,252
CWI (condition 3)				
-Uncorrected errors	,330*	,242	,316	,287
CWI (condition 4)				
-Time (sec)	,438**	,310	,268	,475**

NOTE: Scores used for the analyses were raw scores, except scores from BRIEF-A which were standardized T-scores. BRI= Behavior regulation index, TMT = Trail Making Test, CWI = Color-Word Interference test, * $p<0.05$ ** $p<0.01$, N=38

Table 4. *Correlations between Metacognition Index subscales and neuropsychological tests*

Variable	Metacognition Index Subscales				
	Initiate	Working memory	Plan Organize	Task Monitor	Organisation of Materials
Tower					
-Total achievement score	-,082	-,195	-,183	,004	-,113
TMT (condition 4)					
-Time (sec)	,265	,186	,322*	,405*	,179
CWI (condition 3)					
-Uncorrected errors	-,055	,136	-,028	,126	,230
CWI (condition 4)					
-Time (sec)	,156	,373*	,380*	,332*	,365*

NOTE: Scores used for the analyses were raw scores, except scores from BRIEF-A, which were standardized T-scores. MI= Metacognition index, TMT = Trail Making Test, CWI = Color-Word Interference test, * $p < 0.05$ ** $p < 0.01$, N=38

From Table 3 it becomes clear that the BRI scale Inhibit correlates significantly with time spent on TMT (condition 4) and CWI (condition 4), as well as CWI (condition 3), uncorrected errors. Shift has a significant positive relation to Tower total achievement score and total time spent on TMT (condition 4). Self Monitoring has a strong significant correlation with time spent CWI (condition 4). Therefore, Shift, Inhibit and Self Monitoring are relevant subscales, possibly catching the same executive function as needed to perform CWI (condition 3 and 4), TMT (condition 4) and Tower test. The subscale Emotional Control seems to have no significant relationship with either of the neuropsychological tests. Table 4 illustrates which one of the subscales in MI might tap the same executive functions as the neuropsychological tests. It is apparent from Table 4 that there are few strong relationships between MI and the neuropsychological tests used. All subscales but Initiate have a significant positive correlation with time spent on CWI (condition 4). Only Plan/Organize and Task Monitor have a significant positive correlation with TMT (condition 4) time.

3.4 Relation between BRIEF-A, neuropsychological tests and HSCL-25

The participants in the present study show slightly elevated scores on the depression index of HSCL-25, but not on the anxiety index. As a consequence, the further analyses focus on the relation between BRIEF-A, neuropsychological tests and HSCL-25 depression. Also, the relationship between the Hotel task, neuropsychological tests and HSCL-25 was not analyzed because the Hotel task and HSCL-25 was found to have no significant relationships between them. Bivariate correlation analyses show that the BRIEF-A index BRI has a strong positive correlation to the depression scale of HSCL-25 (0.720, $p \leq 0.01$). The MI index does not correlate with HSCL-25 depression. Consequently, this could possibly confound the relationship between BRIEF-A and the neuropsychological tests, like hypothesized in objective 3. Therefore, partial correlational analyses was performed to shed light on the relationship between BRIEF-A, neuropsychological tests and the HSCL-25 depression scale. The neuropsychological test results selected for this analysis are the ones previously seen to have the highest correlations with BRIEF-A. The results are displayed in Table 5. As seen in Table 5, controlling for HSCL25 depression when analyzing the relationship between BRIEF-A and neuropsychological tests, the correlations has changed compared to correlations between the same tests, as displayed in Table 2, where HSCL-25 depression was not controlled for. None of the previously significant relationships between BRIEF-A and the neuropsychological tests are found.

Table 5. *Partial correlations between BRIEF-A and neuropsychological tests controlled for HSCL-25 depression*

		BRIEF-A		
Control Variables		Global Executive Composite	Metacognition Index	Behavioral Regulation Index
HSCL-25 Depression	Tower			
	- Total achievement score	,062	,031	,070
	TMT (condition 4)			
	- Time (sec)	,247	,285	,160
	CWI (condition 3)			
	- Uncorrected errors	,112	,001	,245
	CWI (condition 4)			
	- Time (sec)	,260	,312	,136

NOTE: Scores used for the analyses were raw scores, except scores from BRIEF-A and HSCL-25, which were standardized T-scores. BRIEF-A = Behavior Regulation Index of Executive Function – Adult version, HSCL25 = Hopkins Symptoms Checklist 25, TMT = Trail Making Test, CWI = Color-Word Interference test, * $p < 0.05$ ** $p < 0.01$, N=38

3.5 Relationship between the Hotel task and neuropsychological tests

Correlational analyses were also performed in order to examine the relationship between the performance based test and the Hotel task (objective 3). As seen in Table 6, there are several positive significant relationships of moderate strength between the Hotel task and the neuropsychological tests chosen for this analysis. The results are in the expected direction, however, the relationships are stronger than predicted. This indicates that the Hotel task to some degree captures the same as the neuropsychological tests, although the size of the bivariate correlations indicate that the two types of tests are not identical.

Table 6. *Correlations between the Hotel task and neuropsychological tests*

Variable	Hotel task			
	Total attempted tasks	Total time deviation	Garage doors	Total time deviation garage doors
Tower				
- Total achievement score	,385*	-,329*	,470**	-,460**
TMT (condition 4)				
- Raw time (sec)	-,457**	,316	-,510**	,527**
- Set loss errors	-,364*	,339*	-,079	,108
CWI (condition 3)				
- Time (sec)	-,495**	,405*	-,478**	,516**
- Uncorrected errors	-,079	,088	-,206	,205
- Corrected errors	-,156	,225	-,082	,083
- Total errors	-,188	,256	-,202	,202
CWI (condition 4)				
- Time (sec)	-,333*	,290	-,424**	,437**
- Uncorrected errors	-,313	,329*	-,338*	,326*
- Corrected errors	-,432**	,441**	-,435**	,397*
- Total errors	-,416**	,435**	-,436**	,411*
CPT				
- Omission	-,243	,193	-,287	,273
- Commission	-,440**	,368*	,013	,008
- Detectability	,491**	-,401*	,106	-,111
- Perseverance	-,390*	,317	-,201	,214

NOTE: Scores used for the analyses were raw scores, CPT = Conners Continuous Performance test II, TMT = Trail Making Test, CWI = Color-Word Interference test. * $p < 0.05$ ** $p < 0.01$, N=38

Stepwise multiple regression analyses were also used to examine how much of the variance in the four Hotel task measures the various neuropsychological tests accounted for taken together. The results of the analyses showed that the neuropsychological tests accounted for 26, 20, 29 and 32 % of the variance in Total attempted tasks, Total time deviation, Garage

doors and Total time deviation garage doors, respectively. This further strengthens the impression of the Hotel task capturing something unique not accounted for by the various the neuropsychological tests.

3.6 Relationship between the Hotel task and BRIEF-A

In order to examine the relationship between the Hotel task and BRIEF-A (objective 4), correlations were calculated between all the subscores of the Hotel task and the main indexes of BRIEF-A. No significant relationships were found. When all subscales from the Hotel task and all the subscales from BRIEF-A were included in the correlation analyses, still no significant relationship were found. The results of these analyses are therefore not presented. The lack of significant relationships between BRIEF-A and the Hotel task suggests that they measure different constructs of executive function. Calculated correlations between the most relevant scores are presented in Table 7.

Table 7. *Correlation between the Hotel task and BRIEF-A*

Variable	Hotel task		
	Attempted tasks	Total time deviation	Total time deviation garage doors
BRIEF-A			
- Global Executive Composite	-0,084	0,040	0,130
- Metacognition Index	-0,048	-0,010	0,150
- Behavioral Regulation Index	-0,120	0,100	0,094

NOTE: Scores used for the analyses were raw scores for the Hotel task and standardized T-scores on BRIEF-A

* $p < 0.05$ ** $p < 0.01$, N=38

4 DISCUSSION

The purpose of this study was to investigate how comprehensive assessment of executive function in adults with SB can provide information of executive dysfunction in accordance to ICF.

The first hypothesis of the current study was that BRIEF-A and neuropsychological measures of executive function does not tap the same level of executive function in line with the ICF model. Previous research on children and adolescents with traumatic brain injury has found weak correlations between performance on neuropsychological tests and executive functions in daily life as measured by BRIEF (Anderson et al., 2002; Vriezen & Pigott, 2002). This could indicate that BRIEF and neuropsychological tests tap different levels of the ICF model. We are unaware of studies examining the abovementioned relationship between BRIEF-A and neuropsychological measures in adults with SB. Hence, in order to investigate whether BRIEF-A can provide additional information on a patient's level of functioning in line with the ICF model, the relationship between BRIEF-A, assessment at the activity level, and neuropsychological tests at the impairment level, was analyzed. The results showed only a few significant correlations between BRIEF-A and the neuropsychological tests. Previous research on the relationship between neuropsychological tests and BRIEF-A on other patient groups have also revealed few correlations between the tests. As a consequence, it was predicted that this holds true in a study of adult SB patients. We hypothesized that if there were few correlations to be found, this could mean that the assessments employed tap different levels of executive function, namely at the impairment level and activity level according to ICF. This relationship supports the argument that BRIEF-A adds information to neuropsychological tests. The analyses of the relationship between BRIEF-A and neuropsychological tests found some moderate correlations, although there were many subscales in BRIEF-A that demonstrated low correspondence with neuropsychological tests, such as Emotional Control, Initiate, Working Memory and Organization of Materials. This indicates that neuropsychological tests and BRIEF-A in the SB patient group both tap some of the same functionality levels.

The second hypothesis of the present study was that an elevated level of psychological distress, as measured by HSCL-25, would affect how executive dysfunction is reflected through BRIEF-A and neuropsychological measures. We believe that no studies have investigated the abovementioned relationship in adults with SB. Partial correlation analyses

were done to investigate the relationship between BRIEF-A and neuropsychological tests, controlling for HSCL-25 depression scale. The results were as expected and the previously documented significant correlations between BRIEF-A and neuropsychological tests were not found, suggesting that depression affects the responses on one of the tests, most likely the BRIEF-A. Controlling for HSCL-25 depression makes the low correspondence between BRIEF-A and neuropsychological tests even clearer.

In cases of low correspondence between self-report questionnaires and neuropsychological tests, previous research indicates that the relationship between subjective and objective measures could be confounded by psychological factors such as symptoms of depression (Julian, et al., 2007; Maor, et al., 2001) or coping mechanisms (Maor, et al., 2001). These factors are mostly related to overestimation of cognitive dysfunction on subjective measures (Julian, et al., 2007). Overestimation of cognitive performance is suggested to relate to less depression, conscientiousness and greater degree of cognitive impairment (Carone et al., 2005). Coping mechanisms is mentioned as a possible factor responsible for the discrepancy between subjective and objective measures of cognitive function. Gold et al. (2003) postulated that negative and positive coping mechanisms (denial, lack of insight or overestimation), and not depression, could be responsible for the discrepancy. The participants in our study showed somewhat elevated scores on the depression index in HSCL-25. This could have affected the scores on BRIEF-A, hence affecting the relationship between BRIEF-A and the neuropsychological tests. It is possible that BRIEF-A taps psychosocial stress on a general level. Since the participants in the present study report symptoms of depression on HSCL-25, this is worth a consideration when interpreting the results. In terms of a comprehensive assessment of executive function with the goal of planning and realizing rehabilitation, it would be important to know whether subjectively reported executive dysfunction is related to brain damage or to depression. This would imply a different rehabilitation approach. There is a dearth of research looking into the effect of psychosocial stress on the accuracy of self-reported questionnaires on behavioral executive dysfunction, and this is an important area to look into in the future.

When a patient experiences severe symptoms of depression, this could also affect neuropsychological tests, especially the timed tests. However, the participants in this study score within the normal range on measures of psychomotor speed such as the TMT and CWI. In conclusion, the low correspondence between neuropsychological tests and BRIEF-A in the

present study suggest that the measures tap different levels of executive dysfunctions. However, this could also mean that BRIEF-A is more sensitive to psychosocial stress in general, and not necessarily to executive function. Nevertheless, the level of psychosocial stress as experienced in daily life is relevant when mapping a patient's function according to the ICF.

The third hypothesis of the current study was that the Hotel task and neuropsychological measures of executive function does not tap the same level of executive function in line with the ICF model. Previous research on other patient groups finds only weak positive correlations between the Hotel task and neuropsychological measures (Torralva et al., 2012). Also, in line with the SAS model as proposed by Norman and Schallice (1986), and its extension (Burgess, 2000; Burgess et al., 2000), there is reason to believe that the Hotel task and neuropsychological measures could relate to different parts of executive function. Consequently, we expected low to moderate correspondence between the Hotel task and neuropsychological tests. The present study found several low to moderate significant positive correlations between the Hotel task and neuropsychological tests employed in this study. This partially supports the hypothesis of the study i.e. that the Hotel task on the activity level taps some of the same areas assessed by neuropsychological measures on the impairment level. However, since the measures were not highly correlated, an implication of the study is that the Hotel task should be included in a comprehensive assessment of measuring executive function in adults with SB. This could especially be true when assessing patients with a subtle executive dysfunction. As seen in Torralva et al's (2009) study, the Hotel task was able to tap executive dysfunction in frontotemporal dementia patients with a high score on Addenbrooke's Cognitive Examination. These patients scored significantly worse than the healthy control group on the Hotel task. At the same time, they did not score differently from healthy controls on classical neuropsychological tests. However, the patients with a low score on ACE, scored significantly worse on both the Hotel task and the neuropsychological tests compared to controls. Torralva et al's (2009) study epitomizes a challenge in the neuropsychological study of executive dysfunction, namely a fractionation of executive symptoms, as suggested by Chan et al. (2008). This delineate the possibility that neuropsychological tests and more life-like measures tap different levels of executive function in accordance to the ICF model, hence making an argument that measurements such as the Hotel task should be considered for inclusion in a comprehensive assessment of executive function.

The fourth hypothesis of this study was that the Hotel task and BRIEF-A does not tap the same level of executive function in line with the ICF model. No other studies than the current one has evaluated the correspondence between the Hotel task and BRIEF-A. According to Chan et al., (2008), the Hotel task is likely to tap the activity level of ICF, whereas BRIEF-A taps both activity and participation level. Therefore, we expected that the Hotel task and BRIEF-A would show few significant correlations. No significant correlations between BRIEF-A and the Hotel task were found, and this supports our hypothesis. This finding is of interest since both BRIEF-A and the Hotel task correlates with neuropsychological tests, although differently, probably due to the fact that they tap different parts of executive functioning. While the Hotel task assesses multitasking abilities in line with the extended version of the SAS model, BRIEF-A is probably more sensitive to psychosocial stress in the environment, thus incorporating how stress affects executive function in daily life. The results delineate the problematic nature of assessing executive dysfunction as well as the controversial term executive function. Another explanation of the observed result could be that BRIEF-A is a self-report questionnaire developed with ecological validity in mind, namely measuring executive functioning in daily life (Gioia & Isquith, 2004)., whereas the Hotel task is a laboratory based test. The Hotel task also aims to predict performance in daily life (Torralva et al., 2009), yet despite this, the Hotel task is laboratory based and still suffers under the same challenges of ecological validity as neuropsychological tests. In a laboratory based environment, neuropsychological tests as well as the Hotel task are administered without disturbances and distractions, unlike the natural environment BRIEF-A addresses. In a natural, daily life environment, the patient is not protected against distractions, and this is one of the most problematic areas of assessment of executive functioning (Lezak, 1982).

The homogeneity of the participants in the current study is worthy of consideration for future research. As seen in Table 1. Descriptive statistics of neuropsychological data, the participants scored mainly below the area considered within the normal range. As a consequence, when measuring patients' with these scores, ecological validity is not the main issue, because most neuropsychological tests would be able to tap this level of executive function. In cases of more subtle executive dysfunction, where neuropsychological assessment fail to pick up an executive deficit, ecological assessment would be of profound importance. Patients with SB are a highly heterogeneous group, and it is likely that a subgroup would score within the normal range on classical neuropsychological tests and display subtle executive dysfunctions. In this setting, the issue of ecological validity becomes

relevant, and the use of comprehensive assessment of executive functions even more important. Future research should address the need for comprehensive assessment in patients with subtle executive dysfunction, as possibly not measured well by classical neuropsychological tests.

However, the results from the current study suggest that BRIEF-A and the Hotel task tap different levels of executive function, hence it supports the suggestion that both the Hotel task and BRIEF-A should be considered for inclusion of a comprehensive assessment of executive function in adults with SB.

4.1 Strengths and limitations of the study

This study is limited by a relatively low sample size ($N=38$), and consequently it is difficult to generalize the findings to all patients with SB, as well as other patient groups. Also, the SB patient group is a heterogeneous group and this has further implications on generalizability.

General limitations of self-report questionnaires such as BRIEF-A is worth noting. A self-report questionnaire has limitations such as the validity of responses. When using a self-report questionnaire the participant's responses must be validated. In this study, validity indexes of Negativity and Inconsistency on the BRIEF-A were acceptable. However, even when a participant answers honestly, there could be lack of insight in terms of under- or over-reporting of symptoms. Prior studies on patients with multiple sclerosis (MS) have investigated the accuracy of self-report questionnaires, and questions of the correspondence between subjective and objective measures have been raised. Some studies have shown that subjective and objective measures, such as self-report questionnaires and neuropsychological tests, coincide in their results (Basso et al., 2008; Marrie et al., 2005), hence they show concurrent validity. In other studies however, the same relationship between subjective and objective measures were not found, and the results of self-report questionnaires and neuropsychological tests do not correlate (Benedict et al., 2003; Middleton et al., 2006). Despite this, we need to keep in mind that a self-report questionnaire like BRIEF-A is asking for the respondents own experiences of a problem, not how a family member or a friend experiences the challenges in daily life (except in an informant form addressing someone close to the patient). According to a close relative or staff at the hospital the patient might be over- or underreporting executive dysfunction and a self-report questionnaire cannot escape this challenge. Also, results on neuropsychological tests could indicate another status of

executive function than self-report measures. However, when asking for the patient's own experience, it is highly probable this will be the answer we get. On the other hand, when assessing executive dysfunction with a self-report questionnaire, level of self-awareness could affect the validity of the measure.

An informant questionnaire is available for BRIEF-A, asking someone close to a patient to answer the same questions as the patient about his/her behavior. Manchester et al. (2004) argues that collateral information would help circumvent the problems many patients with executive dysfunction have with accurately reporting their difficulties. Awareness is an aspect closely related to executive functions, such as planning, self-monitoring and behavioral control (Stuss, 1991). The current study did not make use of the informant form, as the collateral information was not available at the time of the study. The information from the informant questionnaire could have been compared with the participants own answers to analyze whether there were notable discrepancies. In addition, it could have alleviated the abovementioned problem of self-reporting. It is worth noting that collateral information could be of questionable reliability. When collecting informant questionnaires, the participants are asked to choose a person who knows them well. The definition of knowing someone well is relative, and some of the participants may choose someone who is less likely to know how they function on a day-to-day basis. For instance, this could be a friend who visits once or twice a year. One study found that a relative's familiarity with the patient's everyday functioning can affect the accuracy of informant reports (Norris & Tate, 2000). However, this problem could be alleviated by giving clear instructions as to who should perform as a collateral. It is hypothesized that reports from a close one that spends a large amount of time with the patient would offer valid information on the real life difficulties experienced. However, several factors may affect the accuracy of such reports. Family stress and dynamics (Fleming et al., 1996), time since injury (Chevignard et al., 2000), and effects of stereotypical views of patients performance held by some clinicians (Cavallo et al., 1992) are all factors suggested in the literature. There is also evidence pointing to family and staff perceptions being somewhat rigid and may not change even when behavioral difficulties later improve (Sohlberg et al., 1998).

Despite the abovementioned challenges relating to self-reporting instruments like BRIEF-A and collateral information, the current study has been able answer to the questions put forth by the authors on how to perform comprehensive assessment of executive function in adults

with SB. Previous research has been able to demonstrate that children with SB show anatomical abnormalities in the brain, and this is associated with lower scores on intelligence measures (Bier et al., 1997). Several studies have shown that children with SB especially struggle with tasks involving executive dysfunction (Burmeister et al., 2005; Rose & Holmbeck, 2007), and that this is related to psychological adjustment (Kelly et al., 2011). However, there is a dearth of research looking into executive dysfunction in adults with SB, and the measurement of these. We are unaware of studies other than the current, examining how the Hotel task and BRIEF-A could be included in a comprehensive assessment of executive function, in line with the ICF model. Executive dysfunction may cause a number of behavioral difficulties, for instance dealing with novel situations, problems forming a reasonable plan and increased distractibility (Levine et al., 2007). Studies of adolescents with SB indicate that this patient group is likely to experience challenges related to academic life, at work, in social relationships, thus restricting them from participation from society (Barf et al., 2010; Barf et al. 2009; Friedman et al., 2009; Heffelfinger et al., 2008; Hetherington et al., 2006; Holmbeck et al. 2010; Stubberud & Riemer, 2011). The challenges adults face might be even more pervasive. Hence, research into comprehensive assessment of executive functions in adults with SB is of profound significance. This would further give implications on the focus of rehabilitation for the patient as an individual, keeping in mind that the SB patient group is highly heterogeneous.

4.2 Conclusion

The results from this study have demonstrated that BRIEF-A and the Hotel task could provide important information on a patient's level of participation and activity in line with the ICF model, not only the impairment level made accessible through neuropsychological tests. This suggests that BRIEF-A and the Hotel task are measures that should be considered for inclusion when planning a comprehensive assessment of executive function with the goal of measuring levels of ICF. When assessing with the goal of classifying a patient's different levels of functioning, it could ultimately provide an information framework from which to work on a rehabilitation plan.

The participants in the current study displayed an elevated level of symptoms of depression, as measured by HSCL-25. This affected the relationship between BRIEF-A and the neuropsychological tests. In a rehabilitation setting it is of profound importance to understand

whether results on neuropsychological tests are a reflection of psychosocial stress or organic lesion or disease, as this would have implications on the focus of rehabilitation and how it is planned and executed. Therefore, as part of a comprehensive assessment of executive function, it would be paramount to conduct measurements of depression in patients who experience difficulties in their daily life, yet score within the normal range on neuropsychological tests.

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